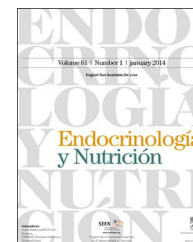




ENDOCRINOLOGÍA Y NUTRICIÓN

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SCIENTIFIC LETTER

Spontaneous neck hematoma due to rupture of parathyroid adenoma: Report of 3 cases and literature review[☆]

Hematoma cervical espontáneo por rotura de un adenoma de paratiroides: 3 casos y revisión de la literatura

Spontaneous hemorrhage from a parathyroid adenoma is an extremely rare but potentially serious complication.^{1,2} Clinical signs and symptoms depend on the volume and location of the hematoma and include a mass in the neck, ecchymosis, pain, and symptoms from the compression of adjacent structures. In extreme cases, the hemorrhage may extend to the mediastinum and the pleural cavity, causing chest pain, cough, and respiratory failure. Imaging tests are helpful to guide diagnosis.³ Both computed tomography (CT) and magnetic resonance imaging (MRI) allow for identifying the anatomical structures and the extent of the bleeding. Differential diagnosis should be made with thyroid gland lesions, superior vena cava syndrome, aortic dissection, or complication of a mediastinal lesion.⁴ Urgent treatment with evacuation of cervical hematoma may be required when airway involvement, hypercalcemic crisis, or active bleeding exist.² The aim of this study was to review our cases to clarify the prevalence and treatment of this complication.

Three cases of primary hyperparathyroidism which appeared as spontaneous neck hematoma recorded in the prospective database of 560 parathyroidectomies from the endocrine surgery unit of our institution were collected. The first case was a 56-year-old woman with a history of high blood pressure and cholecystectomy. She attended the emergency room for sudden pain in the anterior neck region, and reported no prior trauma or anticoagulant or anti-inflammatory treatment. Physical examination revealed blood pressure levels of 190/120 mmHg and ecchymosis in the anterior neck area. Supplemental tests showed hypercalcemia (calcium level, 10.9 mg/dL) and a parathormone (PTH) level of 145 pg/mL. A CT scan of the neck and chest showed diffuse occupation of the left anterior and posterior neck by a hematoma. A tubular lesion

taking up contrast was seen at the left paraesophageal level. No thyroid gland changes or vascular abnormalities were found. A Tc-sestamibi scan showed posterior hyperuptake at the lower pole of the left thyroid lobe. Bilateral parathyroid exploration conducted 3 months later identified an adenoma in the left superior parathyroid gland in a low paraesophageal position.

The second case was a 39-year-old woman who reported neck pain followed 24 h later by a hematoma in the anterior neck and upper chest area. Physical examination revealed an enlarged thyroid gland of normal consistency with no nodules. Thyroid or parathyroid gland bleeding was suspected. Calcium and PTH levels were 11.1 mg/dL and 199 pg/mL respectively. Single photon emission computerized tomography (SPECT-CT) of the parathyroid glands showed left prevertebral hyperuptake behind the left thyroid lobe. Bilateral parathyroid exploration conducted 8 months later identified an adenoma in the left superior parathyroid gland in a low paraesophageal position.

The third case was a 53-year-old woman diagnosed with primary hyperparathyroidism after reporting musculoskeletal pain. Six months earlier, she had had a spontaneous neck hematoma and dysphonia which resolved with no need for medical advice. Calcium and PTH levels were 10.7 mg/dL and 160 pg/mL respectively. Imaging tests were inconclusive. Bilateral cervical exploration found an orthotopic adenoma in the left superior parathyroid gland surrounded by an inflammatory pseudocapsule adhering to adjacent structures.

The first case of cervical and mediastinal hemorrhage from a parathyroid gland lesion was reported by Capps in 1934.¹ Approximately 30 cases have been reported to date in the medical literature. Of these, 86%, 10%, and 3.5% occurred in patients with adenoma, parathyroid hyperplasia, and cyst respectively. The most common signs included dysphagia, dyspnea, pain, and neck ecchymosis.^{2,3} Spontaneous hemorrhage from a parathyroid adenoma is an extremely rare complication (0.4% prevalence in our unit). What causes the spontaneous rupture of a parathyroid adenoma is not known. However, it is thought to be due to a mechanism similar to apoplexy of other endocrine glands. It may therefore be due to an imbalance between gland growth and blood supply which leads to infarction (with necrosis and hemorrhage) that may extend outside the capsule.³ Predisposing factors include trauma, anticoagulant therapy, and treatment with non-steroidal anti-inflammatory drugs.² Parathyroid glands have no preference for any gland, and no greater frequency in

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any specific location has been seen in cases reported in the specialized literature. It should be noted, however, that in all three cases reported here the lesion occupied the left superior parathyroid gland, and in two of them adenomas descending toward the posterior mediastinum were found.

Simic et al. proposed three diagnostic criteria which are still valid: acute neck swelling, hypercalcemia, and neck and chest ecchymosis.⁴ The clinical signs and symptoms depend on hematoma volume and location, and potentially serious complications include airway and esophageal obstruction and hypovolemia.^{5,6} The most common signs and symptoms include neck mass or swelling, hematoma or ecchymosis, pain, dysphagia, dysphonia, or dyspnea due to compression of the esophagus, laryngeal nerve or trachea respectively. Hypercalcemia is common at the time of hemorrhage, but hypocalcemia may be seen if extensive gland destruction occurs. Cases of the spontaneous remission of hyperparathyroidism due to infarction or hemorrhagic necrosis of adenoma have been reported.⁷

Initial X-ray studies are helpful in guiding diagnosis. They usually show tracheal deviation, esophageal compression, or mediastinal widening.⁵ Both CT and MRI allow for the identification of the anatomical structures and the extent of the bleeding, and for ruling out thyroid lesions. If the diagnosis is suspected, work-up must be completed with more specific imaging tests such as a Tc-sestamibi scan.⁸

Diagnosis may be delayed due to non-recognition of the lesion. A high index of suspicion is therefore needed in all patients with unexplained acute neck pain accompanied by inflammation, dyspnea, dysphagia, and dysphonia. In differential diagnosis, thyroid lesions and mediastinal or vascular complications such as aortic dissection or vena cava syndrome, amongst other conditions, should be ruled out.^{2,8}

In stable patients with uncomplicated hematoma, initial conservative management is recommended, and elective surgery with complete cervical exploration should be programmed. By contrast, urgent surgical treatment with evacuation of the hematoma is required when airway involvement, hypercalcemic crisis, or active bleeding occurs.^{2,8,9}

In conclusion, spontaneous rupture and extracapsular hemorrhage from a parathyroid hematoma, although rare, may be serious complications. It should therefore be suspected in all patients with neck hematoma with no history

of trauma. Elevated plasma calcium and PTH levels and decreased hemoglobin values are important for making a final diagnosis.

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